NEUROORTHOPEDIC APPROACH TO TREATING A PATIENT WITH SCHWANNOMA AND AGGRESSIVE HEMANGIOMA AT A SINGLE SPINAL MOTION SEGMENT: A CASE STUDY

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A clinical case of surgical treatment of a female patient with dumbbell schwannoma growing from the T4 nerve root and aggressive hemangioma of the T4 vertebral body and arch is presented. The open kyphoplasty through posterior approach was performed and followed by T4 laminectomy, costotransversectomy at the T4—T5 level on the left, total microsurgical resection of the tumor through right extrapleural approach, and transpedicular fixation of the T3—T5 vertebrae. There were no postoperative complications. The control computer tomography confirmed the complete removal of the tumor and the spine stability at the level of surgery.

Key Words: dumbbell spinal tumor, aggressive hemangioma, schwannoma, kyphoplasty, laminectomy.

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The incidence of primary tumors is 12–14 cases per 100,000 people a year. Moreover, a significant increase in tumor diseases of both the musculo-skeletal system and the central nervous system has been noted for the first time [1].

Dumbbell schwannomas account for about 18 % of tumors of the central nervous system. The name is given due to the unique morphology of the intracanal and paravertebral tumor components. As a rule, these neoplasms are diagnosed quite late, manifesting themselves as symptoms of spinal cord compression. Only timely intrascopic diagnostics enables neoplasm detection at early stages. Such tumors often have a more pronounced paravertebral component [2]. Aggressive bone-destructive growth of the extracanal component sometimes leads to destruction of the body and articular processes of a vertebra. Most often, schwannoma affects cervical spine (44 %), followed by lumbosacral spine (29 %) and thoracic spine (27 %) [3].

Hemangioma is the most common among primary vertebral tumors. Independent morphological studies have shown that hemangiomas of the vertebral bodies are found in the population with rate up to 10.7–11.9 % [4]. Hemangiomas develop in all parts of the spine, but most often in the thoracic (60–76 %) and lumbar spine (21–29 %), less often in cervical spine (2–11 %) and sacrococcygeal region (up to 1%) [5].

Surgical treatment of both dumbbell schwannomas and aggressive hemangiomas is currently not difficult and described in many publications. However, no information has been found on the tactics of treatment for a combination of these two pathologies within the same spinal motion segment. This constitutes a ground for presenting this clinical case.

The type of publication is a clinical case description. Study Evidence Level – V (the American Society for Clinical Oncology (ASCO) classification).

The objective is to analyze the literature and to present the results of surgical treatment of a patient with schwannoma and aggressive hemangioma at the level of one spinal motion segment.

Material and Methods

Prior to the surgery, a female patient had MRI and CT to determine the size and severity of bone tissue destruction. The soft-tissue paravertebral component (tumor) and the degree of its invasion relatively to the anatomical structures of the spinal canal were differentiated.

The diagnosis of aggressive vertebral hemangioma was formulated concerning the score for assessing the aggressiveness of hemangiomas [6].

The severity of the pain syndrome was assessed by VAS; the disability degree was determined by the Oswestry Disability Index (ODI). Neurological disorders before and after the surgery were assessed using the ASIA/IMSOP scale.

The individual specifics of the surgery depended on the nature of the lesion revealed during the preoperative examination.

After the surgical intervention, CT scan was repeated to control the volume of tumor resection, the stability of the implanted hardware and the filling the hemangioma zone with bone cement,

and to find any polymethyl methacrylate migration.

The search for scientific publications over the past 10 years was performed in the eLibrary Russian information and analytical portal, the PubMed English-language text database of medical and biological publications and Web of Science online search platform using the key words: shwannoma (910 items), dumbbell spinal tumor (in Russian) (1072 items), aggressive hemangioma (in Russian) (1565 items), dumbbell-shaped spinal tumor (194 items), aggressive hemangioma (in English) (1019 items).

The patient M., female, 60 y.o., considers herself ill for about 4 years, when for no apparent reason she experienced pain in the thoracic spine. Conservative treatment was ineffective. She felt weakness and numbness in the lower extremities that appeared 3 months before the appealing. Examination revealed hemangioma of the body and arch of the T4 vertebra and dumbbell tumor at the level of T4–T5 vertebrae (Fig. 1, 2). She was admitted to the Scientific Research Institute of Traumatology, Orthopedics and Neurosurgery of Saratov State Medical University n.a. V.I. Razumovsky for surgical treatment.

The patient's general condition was satisfactory at the time of examination. Communicative, adequate, oriented. No pathologies were found in the somatic status.

Pain in the thoracic spine was assessed by the patient at 8 points of VAS, the ODI score was 28 points. In the neurological status, lower spastic paraparesis was noted up to 4 points; impaired surface sensitivity of the spinal conduction type from the T6 level on both sides, the kinesthetic sense was lost on the left. According to the ASIA/IMSOP standard, motor status corresponded to type C.

Surgery. Patient was laid in the prone position. An incision was made in the skin and subcutaneous tissue in the plane of the spinous processes of T3–T5 vertebrae. Skeletization and decortication of the posterior supporting structures of the spine were performed. Attention was drawn to the hemangiomatous restructuring of the T4 vertebral arch, profuse bleeding was noted in the course of its skeletization and an attempted resection. A left-sided transpedicular insertion of the trocar to the center of the T4 vertebra body was made under X-ray control; a tunnel for the balloon was made with a drill. A hydraulic balloon was inserted and inflated (Fig. 3). Further, 3.5 ml of bone cement was injected into the vertebral body through the trocar. Laminectomy of the T4 vertebra was performed; the bleeding during arch resection after the insertion of bone cement was minimal. The dural sac was visualized. It was dislocated to the right by an extradural tumor of a densely elastic consistency and gray-cyanotic color (Fig. 4a). Facetectomy, foraminotomy of the T4–T5 on the left were performed. The subperiosteal proximal segments of the 4th and 5th ribs on the left for 5 cm were skeletonized. Costotransversectomy of the T4, T5 on the left was performed. The paravertebral component of the tumor, spreading to posterior mediastinum, was visualized. The intracanal and paravertebral components of the tumor were mobilized with the use of microsurgical techniques. The tumor was supplied abundantly with blood from the paravertebral tissues. Numerous feeding vessels are mobilized, coagulated and transected. The tumor matrix was verified on the left T5 root that was transected at the edge of the dura mater. The tumor was completely removed en bloc (Fig. 4b). Trans-

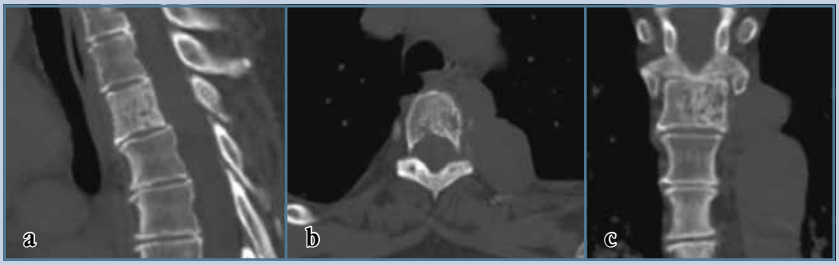

Fig 1
Sagittal **(a)**, axial **(b)** and frontal **(c)** MSCT sections of the thoracic spine in patient M., female, 60 years old, before the surgery: an extradural tumor with a paravertebral left-sided component of the thoracic spine at the level of T4–T5 vertebrae; hemangioma of the body and arch of the T4 vertebra

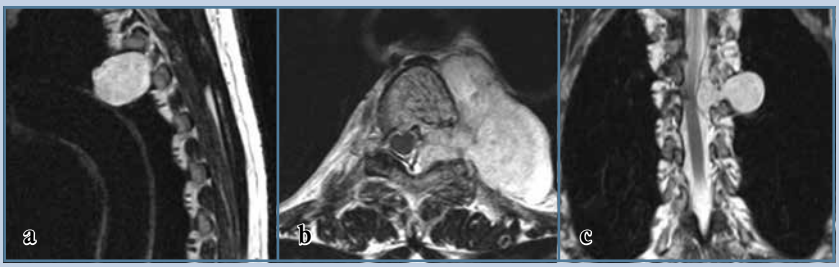

Fig 2
Sagittal **(a)**, axial **(b)** and frontal **(c)** MRI sections of the thoracic spine in patient M., female, 60 years old, before the surgery: an extramedullary extradural neoplasm in the spinal canal with a left-sided paravertebral component at the level of T4–T5 vertebrae, with extension through the region of the left intervertebral dumbbell foramens, total size 5.9 × 4.5 × 2.4 cm, with compression and dislocation of the dural sac to the right; hemangioma of the body and arch of the T4 vertebra with a discontinuous contour of the posterior endplate of the body

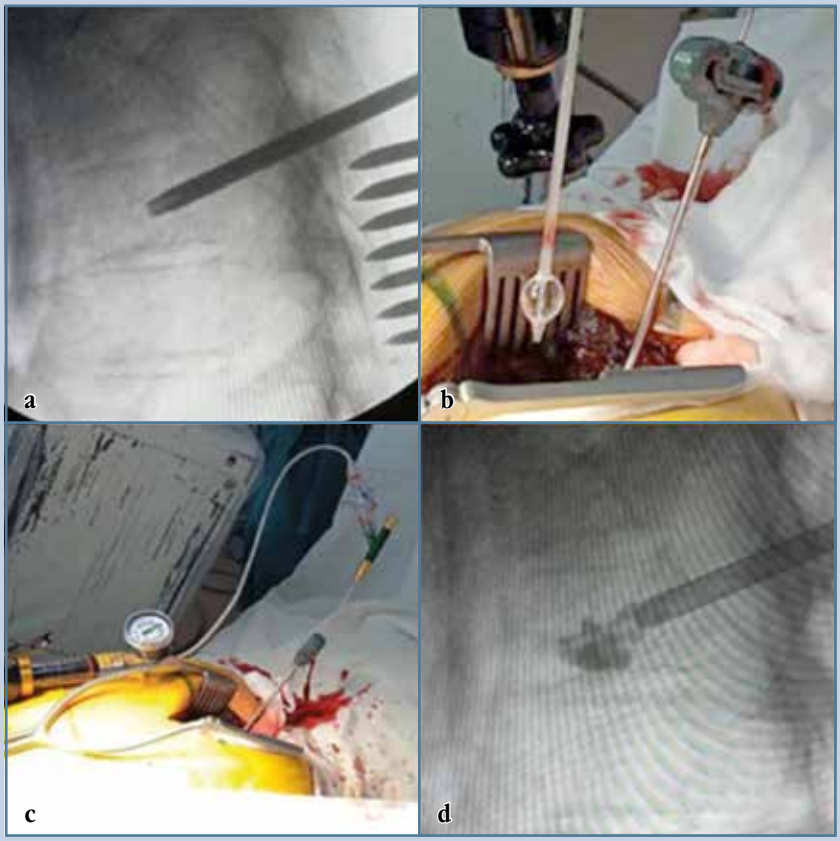

Рис. 3
Open balloon kyphoplasty of the T4 vertebral body (intraoperative image): **a** – a trocar was inserted in the body of the affected vertebra; **b, c** – insertion of a balloon for kyphoplasty; **d** – dilatation of the balloon with contrast solution

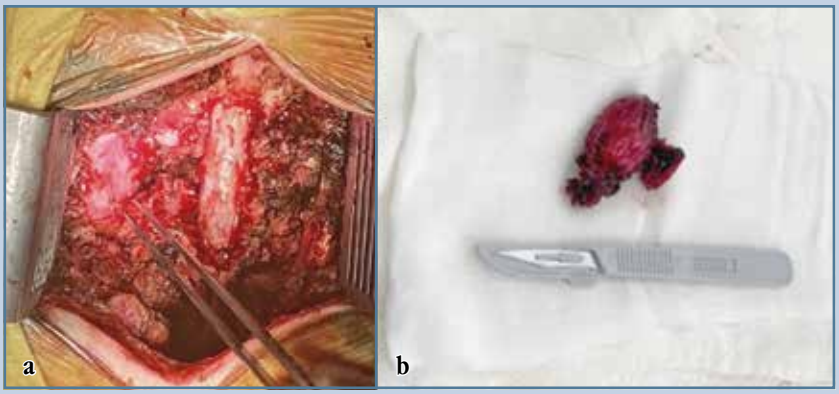

Fig 4
Laminectomy of the T4 vertebra, costotransversectomy of the T4–T5 vertebrae on the left (intraoperative image): **a** – tweezers indicate the paravertebral part of the dumbbell tumor (schwannoma); **b** – gross specimen: dumbbell tumor (schwannoma)

pedicular screws were installed bilaterally in the bodies of the T3 and T5 vertebrae. Then they were connected by two rods and fixed with nuts. A transverse connector is installed to ensure rotational stability. Decortication of the arches of T3 and T5 vertebrae and posterior fusion with synthetic osteoinductive material were performed. Hemostasis. Two active drains were installed in the area of the posterior mediastinum. Layered wound closure. Total intraoperative blood loss was 150 ml.

CT-scan was done on the 1st day after surgery (Fig. 5).

The specimens were identified with the histological study as schwannoma and hemangioma (Fig. 6).

In the postoperative period the pain syndrome intensity decreased to 3 points of VAS, ODI score decreased to 14. Regression of the neurological deficit (E on the ASIA/IMSOP scale) was noted. Full functional recovery was diagnosed 6 months after the surgery (0 by ODI).

Discussion

Surgical intervention for dumbbell schwannoma is indicated for patients with progressive neurological deficit or intrascopic signs of tumor growth. A special growth of neoplasms with development of two parts connected by an isthmus in the intervertebral foramen requires a special approach to their treatment. Not only the intracanal part of the tumor must be removed, but also its paravertebral and extraforaminal components, that are spread into interfascial spaces, mediastinum, pleural cavity and retroperitoneal space [3]. The surgery involves the total removal of the neoplasm in order to minimize the risk of its recurrence. There are options described using both isolated posterior and combined (laminectomy + thoracotomy or thoracoscopic resection of the paravertebral component of the tumor) approaches in case of thoracic spine lesion [7]. The optimal type of surgical approach remains debatable due to the relative rarity of such tumor type.

Indications for surgical treatment of spinal hemangiomas are signs of their

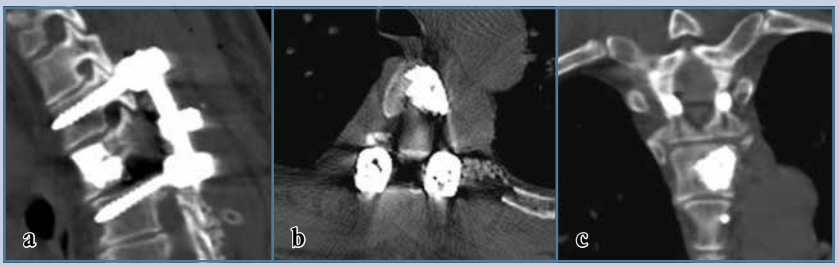

Fig 5
Sagittal **(a)**, axial **(b)** and frontal **(c)** MSCT sections of the thoracic spine in patient M., female, 60 years old, after the surgery: laminectomy of the T4 vertebra, costotransversectomy of the T4–T5 vertebrae on the left, microsurgical complete resection of the tumor at the level of T4–T5 vertebrae, augmentation of the T4 vertebral body with bone cement, transpedicular fixation in T3–T5 segments

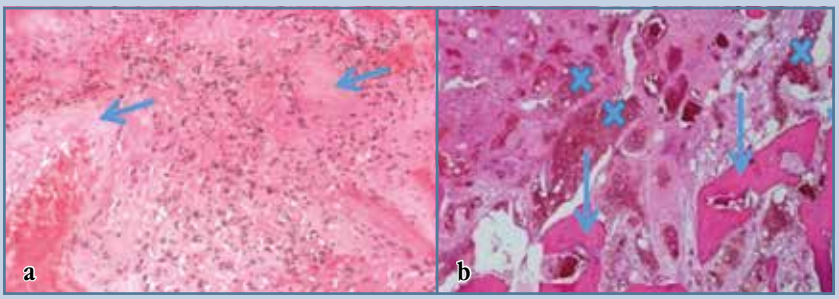

Fig 6
Histological image of schwannoma **(a)** and hemangioma **(b)**, stained with hematoxylin-eosin: **a** – schwannomas is represented by full-blooded and thrombosed vessels with sharply sclerotic walls, including a hyalinized stroma with few clusters of elongated cells, in places forming Verocay bodies (marked with arrows); mag.: lens – 10, eyepiece – 40; **b** – thin-walled blood vessels of various sizes, filled with blood and serous fluid, pass through the bone trabecules of the vertebral body, replacing the normal bone marrow; arrows indicate bone trabecules of the vertebral body, crosses indicate thin-walled blood vessels; mag.: lens – 10, eyepiece – 40

proven aggressiveness: an extravertebral component, a compression fracture or compression deformity of a vertebral body, bone expansion with protrusion of the cortical layer (vertebra swelling), damage (thinning and/or destruction) of a cortical layer, hemangioma affecting more then 1/3 of the vertebral body volume, an uneven trabecular structure, hemangioma expansion to the pedicle of a vertebral arch, absence of adipose tissue in the structure of hemangiomas (a low signal on T1 and a high signal on T2 WI on MRI, a high signal on T2 VI in the fat suppression mode), the local pain syndrome [6, 8]. The extent of surgery is determined by the specifics of a vertebra lesion, and puncture vertebroplasty continues to be considered the gold standard. Total lesion of a vertebral body by aggressive hemangioma, including those with a defect in the cortical layer, is quite common (44 % of cases). In these cases, extravertebral migration of bone cement is often observed when performing puncture vertebroplasty (40.0–87.5 %) [9].

An attempt was made to search for similar clinical cases in order to optimize the tactics of the planned decompressive and stabilizing neuroorthopedic intervention. We found a single case of mentioning both these nosologies in the same publication – a paper by Huang et al. [10], devoted to the description of the surgical treatment of a patient with an extradural dumbbell tumor that, according to cytomorphology, turned out to be not a schwannoma, characterized by such macroscopic structure, but a cavernous hemangioma. This case does not correspond to the specifics of the clinical case presented by us. About 400 papers focus on surgical treatment of schwannomas and other tumors with a similar type of growth as an isolated nosology, and 1800 papers are devoted to surgery of aggressive hemangiomas. Despite a variety of publications, an analysis of literature sources have not revealed any cases of a combination of an dumbbell tumor and an aggressive hemangioma within the same spinal motion segment.

The neuroorthopedic approach to dumbbell tumors, including schwannoma, is described most detailed by Li et al. [11]. Upon removing a tumor, the authors used unilateral monosegmental transpedicular fixation and transforaminal interbody fusion to compensate for impaired spinal stability after hemilaminectomy and facetectomy. Good intrascopic and clinical results were obtained. Decompression and stabilization surgeries for aggressive hemangiomas with intracanal extension are described in detail in Chen et al. [8]. The combination of posterior fixation with vertebroplasty of the affected vertebra is considered as one of the acceptable options for surgical reconstruction.

In the Russian literature, the priority in the tactics of surgical treatment of dumbbell schwannomas is given to microsurgical interventions through the posterior approach, during which intra- and extracanal tumor components are removed [1, 2]. An additional ablastic effect is achieved, among other things, through laser technologies [3].

Several issues were considered during the preoperative planning and surgical intervention:

– the presence of a persistent moderate pain syndrome preceded the develop-

ment of neurological symptoms for four years, with an obtained X-ray pattern, made it possible to consider the hemangioma of the T4 vertebra as both aggressive and symptomatic;

– the choice in favor of balloon kyphoplasty compared with possible vertebroplasty in order to reduce surgical blood loss in aggressive vertebral hemangioma is made due to a defect in the posterior endplate of the vertebral body detected during preoperative examination (Fig. 2b) and, respectively, the risk of intracanal leakage of bone cement;

– embolization of most of the vessel bed of hemangioma with polymethyl methacrylate facilitated visual control of bone resection and enabled to perform complete en bloc removal of large with paravertebral and intracanal extension and to stabilize the affected spine through a single surgical approach.

A good clinical and intrascopic result allows us to consider this tactic as one of the options for surgical treatment of patients with a combination of pathologies.

Conclusion

The combination of two pathologies (hemangioma and schwannoma) at the same level of the spine is apparently rare. Surgical intervention through a single posterior approach including laminectomy, costotransversectomy, filling a hemangioma-affected vertebra with bone cement, and bisegmental transpedicular fixation allowed performing total tumor resection, stabilization of the spinal motion segment, and early patient activation in the immediate postoperative period. This neuroorthopedic intervention can be recommended as one of the surgical treatment options for rare combination of the described nosologies.

The study had no sponsors. The authors declare that they have no conflict of interest.

Ethical expertise. The patient gave her consent to the processing and publication of personal data. The performed therapeutic measures were completely within the framework of the modern surgical tactics. (No. 1 Protocol of the local Ethics Committee of the Research Institute of Traumatology, Orthopedics and Neurosurgery at Saratov State Medical University as of 01.12.2021)

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